

FLEXIBLE INTRAMEDULLARY NAILING IN THE MANAGEMENT OF SIMPLE BONE CYST

**A dissertation submitted to the Tamil Nadu Dr. M.G.R.
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CERTIFICATE

This is to certify that this dissertation titled “FLEXIBLE INTRAMEDULLARY NAILING IN THE MANAGEMENT OF SIMPLE BONE CYST ” is a bonafide work done by Dr KAUSHIK BHOWMICK, in the Department of Orthopaedic Surgery, Christian Medical College and Hospital, Vellore, in partial fulfillment of the rules and regulations of the Tamil Nadu Dr M.G.R. Medical University for the award of M.S. Degree (Branch-II) Orthopaedic Surgery under the supervision and guidance of Prof. VRISHA MADHURI during the period of his post-graduate study from March 2008 to February 2011.

This consolidated report presented herein is based on bonafide cases, studied by the candidate himself

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AIMS

The aim of this study is to evaluate the efficacy and safety of flexible intramedullary nails in the treatment of simple bone cyst supplemented with bone substitutes in children.

OBJECTIVES

1. To define the clinical profile of children with simple bone cyst.
2. To evaluate the outcome of surgery with respect to cyst healing.
3. To evaluate the various complications with this line of management.

INTRODUCTION

Simple bone cysts constitute 2-3% of benign lytic bone lesions in children with preponderance in the first and second decade of life with 70% of patients presenting with pathological fractures. (4, 32, 35) Since its description by Virchow in 1876, the etiopathogenesis and treatment had confounded surgeons till now. Till the 1950's simple bone cyst was managed by masterly inactivity, irradiation, curettage and bone grafting with or without chemical cauterisation. (12, 32, 34) All these techniques were associated with complications such as high rates of recurrence, pathological fracture premature physal closure and infection. (1, 2, 12, 13). In the late 1970's, steroids and bone marrow was introduced in the treatment of simple bone cyst but the results were found to be unpredictable. (46) In the 2000's bone ceramics such as medical grade calcium sulphate and high porosity hydroxyapatite was introduced in the treatment. (22) But all these modes of treatment did not address the problem of postoperative stability because patients continued to be treated with cast immobilisation which continued the morbidity in these patients.

Flexible intramedullary nail was introduced in the 1980's in the treatment of simple bone cyst but it became popular as an option of treatment in the 2000's. The stated advantage of intramedullary nail was that it provided continuous decompression as well as internal splinting in cases of pathological fracture. (26, 27, 28) We have added ceramic scaffolds to the flexible intramedullary nail to hasten the healing time. This retrospective study evaluates our experience with this technique.

HISTORICAL REVIEW

Virchow in 1876 is credited with the first radiological description of a simple bone cyst by many authors. (22,30,43) Bloodgood in 1910 described the pathology of simple bone cyst as a distinct entity from other cystic lesions.(2) They are not true neoplasms but they create structural defect in the long bones like the humerus and femur causing fractures. Patients usually present when they have pain due to pathological fracture or when incidental radiograph shows a defect in the bone.

Jaffe and Lichtenstein traced the natural history of simple bone cyst and suggested that the disease is most active in children below the age of 10 years and becomes inactive after that. They found that a cyst does not progress after closure of the growth plates. About 15% of simple bone cysts undergo spontaneous resolution.(32)

Many theories exist regarding its etiopathogenesis, but the etiology remains elusive.(43)

Treatment has varied since the original description, however, simple bone cysts are known for their fortacity and high rates of recurrence.(26,27)

Alldredge et al in 1942 reported treatment in 152 cases of cystic lesions of the bone with various modalities like irradiation alone, irradiation before or after simple curettage, simple curettage, bone grafting or chemical cauterization, complete resection and amputation. There were many complications associated with irradiation such as stiffness, high rates of fractures of the cystic lesion, physal injury and malignant transformation and thus it is no longer in vogue. There was no

evidence that preoperative or postoperative radiation was of any benefit in these patients. It was also stated that resection of bone is a curative option for nonessential or non weight bearing bones like the fibula.(34)

Neer et al in 1966 showed that curettage and bone grafting with or without cauterisation were associated with recurrence rates of 40%. They made an opening in the cortex, curetting the membranous lining and filling the defect cavity with chips or strips of autogenous or homogenous bone graft. They proved that surgical treatment showed better results than masterly inactivity.(12,32,34) An analysis was done for the conservatively treated cyst with respect to the proximity to the epiphysis. Cysts which were 0.5 cm from the epiphysis were treated conservatively. In their study, 92% of the conservatively treated cases were eventually operated.

Spence et al in 1969 used cancellous freeze dried allograft to pack the cyst cavity after curettage showing a healing rate of 64%. In 1976, Spence et al used crushed cortical bone allograft showing a healing rate of 88%. They also showed that cortical bone is better than cancellous bone for packing the cystic cavity because they are retained in the cyst for a longer period of time providing osteogenic stimulus for new bone formation and healing. (33,35)

Fahey et al in 1973 described subtotal resection of the cystic lesion with bone grafting with recurrence rate of 5%. They described the procedure as resection of part of the cyst wall, curetting the lesion and filling the defect with strut graft from the iliac crest or tibia in the major long bones.(9) McKay et al in 1977 described subtotal resection without bone grafting with recurrence of 9%.(16)

Peltier and Jones in 1978 used curettage and packing of the cyst cavity with sterilized plaster of Paris ($\text{CaSO}_4 \cdot \frac{1}{2}\text{H}_2\text{O}$) pellets obtaining a 90% success (cure)

rate. Dreesmann in 1892 was the first one to use plaster of Paris pellets to fill cavities in the bone. In their study, they listed many advantages of using POP pellets as they are cheap and readily available ,could be easily fabricated into pellets or rods, They are radiopaque, easily absorbed ,have a long shelf life ,easily sterilised and does not give local inflammatory reactions in the body.(21)

Scaglietti et al in 1979 reported 90% favourable results using intracystic injections of methylprednisolone acetate, with no growth inhibition or secondary deformity of the involved bone. When these results were carefully reviewed, a number of treated cysts designated as successes never completely healed radiographically, but were clinically stable and thus were reported as a favourable result.(17)

Campanacci et al produced similar results in another series of patients with 15% recurrence but had incomplete healing in 50% of the reported cures.(14,19)

Schreuder et al in 1997 described curettage and bone grafting with cryosurgery using liquid nitrogen with recurrence rates of 12%. (40)

Drilling alone of these cysts has also been reported as well as trepanation with drainage of the cyst fluid and saline lavage of the cavity. The technique consisted of introducing a trocar to aspirate the cyst fluid and making multiple holes through the cyst wall in all directions. The excretion of cyst fluid is an important step in the mode of treatment.(20)

Lockie et al in 1996 used autologous bone marrow injections for the treatment of simple bone cyst showing comparable results to steroid injections.(19,30).Rougraff et al used demineralised bone matrix in combination with bone marrow showing 22% recurrences after the first injection and no recurrences after the second

injection.(8)Dissatisfaction with steroid injections in some patients, especially those under 10 years of age, has regenerated interest in use of bone graft substitutes. Such substitutes include bioactive ceramics like tricalcium phosphate, calcium sulphate pellets and high porosity hydroxyapatite.(22,24)

Catier et al in 1981 described the use of flexible intramedullary nail in the fixation of pathological fractures in simple bone cyst. Before this, patients with pathological fractures were either treated with a sling or plaster immobilisation and plate screw fixation. He had reported 100% rate of healing in all the patients in his study. Since then, there have been many studies that have shown excellent results with flexible intramedullary nail in children.(26,27,43,44)

LITERATURE REVIEW

Simple bone cyst is a benign, single chambered fluid filled cystic lesion that occurs in the metaphysis of long bones in children recognised as distinctive entity since 1910.

DEMOGRAPHIC CHARACTERISTICS

It is the most common benign lytic lesion in childhood and represents 2-3% of all bone lesions in childhood.(4,26,36)

Incidence is 1 in 10000 cases per year.(4)

Male preponderance exists in the ratio of 2:1 to 3:1(4,32,35)

The lesion is relatively common and usually manifests during the first two decades of life.(32,38).

70% of patients present with pathological fracture.(32)

ANATOMICAL LOCATION

The common location of the cyst tends to be in the proximal end of humerus(75-80%) and the proximal femur(47%).(12,32,35)

Less common locations are pelvis, calcaneum, talus, distal radius, distal femur and proximal tibia.

PATHOGENESIS

There are many theories for formation of the lesion:-

-Virchow's theory-According to Virchow, simple bone cyst is a chondromatous lesion which has undergone cystic softening. This theory was not accepted as there were no facts supporting this theory. (3)

-Monckeberg's theory- it is the healing form of a pre-existing giant cell tumor or osteitisfibrosa. This theory was negated by the fact that giant cell tumors appear at a later age and usually start in the epiphysis whereas simple bone cysts starts in the metaphysis. The histological architecture also does not correspond to simple bone cyst. Strands of giant cells if present are not embedded in the stromal tissue. (1,3)

-Phemister and Gordon's theory - it is secondary to a form of osteomyelitis. Phemister recovered *Streptococcus viridans* from four cases of simple bone cyst but did not yield anything in rest of the cases. The argument against this theory was that the cysts were of long standing duration and may have sterilised themselves. Furthermore, histological studies showed that the lining of these cysts resembled lining of cyst found in the end of long bones in arthritis deformans.(1,13)

-Pommer's theory - there is an encapsulation of a metaphyseal hemorrhage. After the encapsulation, it is kept distended by the transudation of fluid into the cyst and the pressure from the cyst causes stagnation of the lymph and blood vessels resulting in pressure erosion and expansion of the cortex. Objections were raised as patients with fracture who had intramedullary haemorrhage did not develop any cystic

changes. Hence it was postulated that cysts developed in mild trauma without fractures but intramedullary haemorrhage.(1,3,32)

-Mikulicz's theory-he postulated that simple bone cyst represent a local post-traumatic dystrophy. He suggested that the cyst represented some local disturbance in bone growth and development as it had a predilection towards young people and for regions of active growth of long bones. There is microtrauma at the epiphyseal line leading to defect in the enchondral bone formation with subsequent cyst formation.(3)

-Developmental anomaly occurs in the veins of the affected bone with resultant accumulation of interstitial fluid and subsequent equilibration of this fluid with that in unblocked vessels. It was very difficult to prove developmental anomaly in small veins through intraosseous venography. (5,7)

Out of these, the main theories which have gained acceptance are:-

1. Mechanical theory (Cohen's hypothesis)

In the bones of children, the metaphyses are the sites of most rapid remodelling of bone. This is a consistent property of bone with a process of deposition and resorption. When the process of resorption is very rapid, it is accompanied by formation of foci of loose fibrous tissue which resembles that seen in the wall of the cyst. One or more of these foci of fibrous tissue represents the first step in the formation of these cysts.

The vessels present in this area are extremely thin walled sinusoids with extremely slow flow and pressure. Considering the dynamic aspects of flow of interstitial fluid

into the vascular system and the interchange of plasma proteins between the interstitial fluid and plasma, which depends on the lymphatic system, the formation of a bone cyst may be predicted on the blockage in the drainage of interstitial fluid within the metaphysis.

The site of accumulation of this fluid might be well in an area of interstitial tissue where the sinusoidal vessels were occluded by trauma or thrombosis. Another possibility is that one of the branches of the sinusoidal vessels proliferating at the face of epiphyseal cartilage or the rapidly remodelling metaphyseal zone may be partially or completely blocked. Indirect evidence which support this hypothesis is complete interruption of the central medullary vein by intraosseous venography and dilation of the nutrient artery by angiography. On histological examination, congested capillaries and veins are seen in the newly formed cortex and between the spongiform trabeculae near the cyst.

Since the hemodynamic pressure in this area is very low, only a small increase in pressure of the blocked area would be necessary for continuing expansion of the cyst.

The lack of bone trabeculae in the cyst is due to its resorption due to the elevated hydrodynamic pressure in the cyst or altered composition of the fluid stagnating inside the cyst.

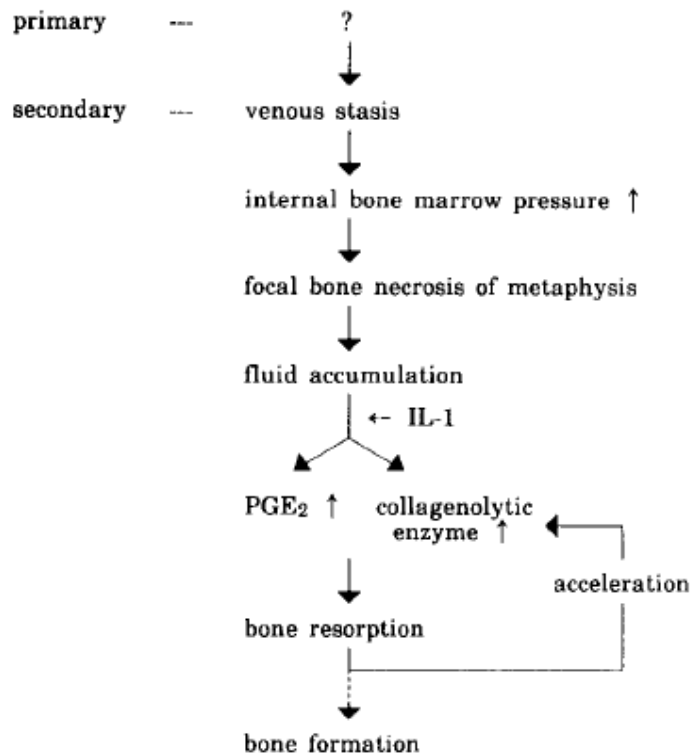
The lining of the cysts are flattened fibroblastic cells which had lead to the earlier speculation of the fluid being formed by either secretory or diffusion mechanism which has been negated now. There is no characteristic resemblance of the cyst fluid with synovial fluid.(7,32)

2. Biological theory (Komiya's hypothesis)

Studies by Komiya et al have shown bone resorptive factors in the cyst fluid such as IL-1, PG and gelatinase. Values of Interleukin- 1β of cyst fluid were examined with an enzyme-linked immunosorbent assay (ELISA) kit. Gelatinase activity in cyst fluid was detected by sodium dodecyl sulfate polyacrylamide gel electrophoresis (SDS-PAGE)

PG E2 is produced by osteoblastic cells in the presence of IL- 1β which induce bone resorption through stimulation of osteoclastic activity. Interleukin-I produced by monocytes or polynuclear cells present in the exudation fluid, stimulates osteoblasts, fibroblasts, or other connective tissue cells to generate PGE 2. IL1 also acts directly on osteoclasts by direct osteoclastic precursor proliferation or maturation. Lymphotoxin, TNF, PDGF and EGF also stimulate bone resorption.

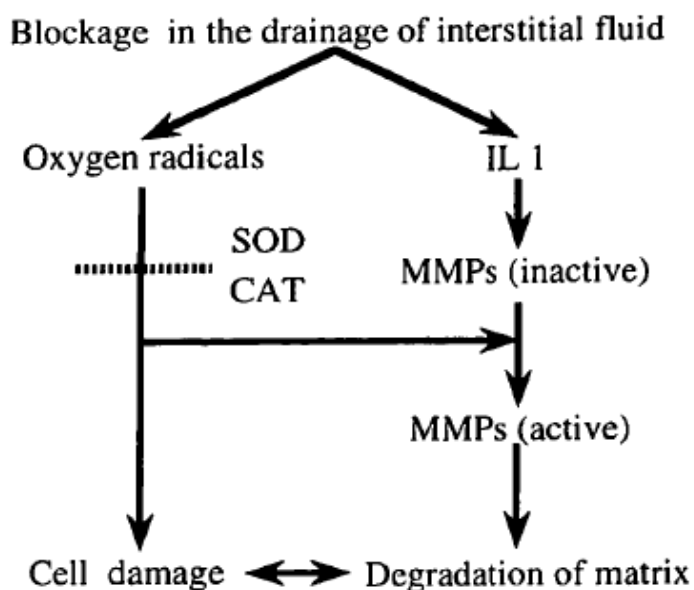
The degradation of the extracellular matrix components of collagens and proteoglycans are attributed to proteolytic enzymes such as collagenase, gelatinase and stromelysin. In the process of bone resorption in a simple bone cyst, gelatinase may play a role in collagenolysis by splitting collagen into two fragments by collagenase.(5)



Komiya et al Oxygen scavengers in simple bone cyst CORR no 287 Feb 1993

Studies have shown high levels of oxygen free radicals in the cyst fluid which contribute to bone destruction. Venous obstruction causes localised ischemia in its drainage field generating oxygen free radicals like superoxide anion(O_2^-), hydrogen peroxide(H_2O_2), hypochlorous acid($HOCl$) and hydroxyl radical(OH) all of which cause cell membrane damage and breakdown of matrix by activating latent collagenase and metalloproteinases. High levels of oxygen scavengers are present in the cyst fluid reflecting the presence of oxygen free radicals as these radicals have a very short half life. These oxygen scavengers are superoxide dismutase and catalase both of which showed increased activity in the cyst fluid.

Superoxide dismutase levels were determined by electron spin resonance spin-trapping technique and the spectra was recorded at 100KHz magnetic field modulation. The spin resonance technique is based on a method for determining superoxide radical (O_2^-). The cyst fluid to be assayed is added in a reaction medium containing hypoxanthine-xanthine oxidase system. Catalase activity is determined by a spectrophotometer by the decomposition of H_2O_2 .(20)



Blockage in the drainage of interstitial fluid leads to ischemia and the generation of oxygen species and IL-1 in the cyst fluid. Oxygen species damage bone cells and degrade bone matrix by activating collagenase and metalloproteinases of latent type. IL-1 also activates metalloproteinases causing matrix breakdown. Oxygen scavengers play a protective role against oxygen species.

Summarising, Cohen's hypothesis is the currently accepted theory of pathogenesis of simple bone cyst.(4,30)Komiya et al have studied the cyst fluid characteristics furthering the pathogenesis of simple bone cyst. They have also supported Cohen's

hypothesis by showing indirect evidence of elevated cyst pressure, dilation of the nutrient artery and interruption of the central medullary vein by intra-osseous venography.(20)

GROSS ANATOMY

Simple bone cysts are found in tubular bones in 90-95% of patients. It usually reaches maximum size before skeletal maturity. Within the long bones, most simple bone cysts are situated in the proximal metaphysis. The involved bone is not wider than the adjacent physis. It is usually a unilocular or rarely multilocular cavity filled lesion with serous or serosanguinous fluid which is yellow or greenish in colour with low viscosity. Inner surface has ridges separating the depressed zone, sometimes the wall is covered by a layer of fleshy tissue of 1cm or more. Occasionally, partial or complete septa are seen, the latter making it multicameral. Periosteal bone formation is slight or usually absent.(7,9,12,38)

CYST FLUID

The serous fluid has been analysed and has been found to have prostaglandins E, Interleukin 1 and metalloproteinases such as gelatinase and collagenase.(5)

HISTOLOGICAL FEATURES

The cyst is lined by a thin connective tissue membrane beneath a thin distended cortex. The cyst is filled with coarse red –brown granulations loosely attached to the lining. Thicker areas when formed are composed of fibrogenic connective tissue that contain numerous benign giant cells, hemosiderin pigment, few chronic inflammatory cells and lipophages.

The granulations are composed of immature fibrous tissue in which are scattered areas of recent and old hemorrhage. The granulation tissue and the lining are extensively infiltrated with lymphocytes plasma cells, polymorphonuclear leucocytes and giant cells of foreign body type. Proliferating fibroblast tissue and callus may be prominent outside and within the cyst.

The bone surrounding the cyst cavity exhibits lacunar resorption alternating with areas of osteoid tissue which may exhibit calcification and new bone formation. Rarely, there are small islets of cartilage also. (12,13,38)

RADIOLOGICAL FEATURES

A. Radiographic features- The typical radiographic appearance is that of a lesion concentrically located in the medullary cavity of the metaphysis of a long bone with expansion in all directions, creating an expanded and thinned but unpenetrated cortex. Cysts located in flat bones such as the pelvis are centred between the inner and outer tables of the ilium.

Cortex is thin and eroded but intact unless pathological fracture has occurred. Fine trabeculations are seen and a healed fracture may be evident as a partition through

it. The long axis of the lesion in the bone almost always exceeds its width, giving the appearance of a truncated cone. The distal extent of the long axis of the cyst, especially in the humerus, is frequently hard to see on plain films and appears to blend into the metadiaphyseal bone outline. If periosteal reaction is present along the thinned cortex of the cyst wall, one should search carefully for a pathologic fracture in the area. Serial radiographs show epiphysis growing away from the region of the cyst so that it lies in the centre of the shaft.(32,38)

The "fallen leaf" sign on plain films is virtually pathognomic of a multiloculated bone cyst. This results when a pathologic fracture in the thinned cortical wall of the bone dislodges a fragment of the cortex. The fragment "drops" into the fluid-containing (but often not filled) cavity of the cyst. This fragment can also shift in the fluid, depending on positioning of the patient's involved bone during serial imaging. The cyst is infrequently distributed from one end of the bone to its opposite end. This appearance is almost always found in the skeletally mature or close to skeletally mature patient.(14, 15)

Pathogenesis of the fallen fragment sign:-

A. Solitary bone cyst in the proximal end of the humerus. Thinned cortex shows "eggshell" cracks following minimal trauma. Periosteum remains intact around the comminuted undisplaced fragments.

B. A large cortical fragment attached to the underlying periosteum only at the bone has become a hinge-like flap (also referred to as a trap door" fragment).

C. Cortical fragment has now become completely displaced and lies at the bottom of the cyst cavity creating the "fallen fragment sign".

D. Lying free in the cyst cavity, the fragment is able to gravitate toward the top of the cavity on change in position of the arm. (15)



Fallen fragment sign.

Radiographs are also used for finding various aspects of simple bone cyst:-

1. Activity

A cyst is considered active if it is

- abutting the physis

- symptomatic with activities of daily living

- has fractured once or more

- has increased in size within an observation period of 3 to 6 months(3,8)

Latent cyst- lesser chance of recurrence. They are further classified as:-

- Latent primary cyst is one in which the distance between the nearest epiphyseal plate and the cyst does not exceed one- third the length of the shaft.

- Latent secondary cyst is one in which the distance between the nearest epiphyseal plate and the cyst exceeds one third the length of the shaft.(9)

2. Size of the cyst- The size of cysts was determined with regard to the length of the affected bone. Cysts involving up to one tenth of the bone length were defined as small, those upto one fifth as medium, and those exceeding one fifth of the bone length as large.(10)

3. Risk of fracture - The fracture risk was evaluated by two methods

Cyst index of Kaelin and MacEwen- One or more trapezoids were drawn round the cyst to measure the cyst area. The diameter of the diaphysis was measured in its tubular part. The cyst index is the cyst area divided by diameter squared. The lower limits of the cyst index for pathologic fracture were 4 for the proximal humerus and 3.5 for the proximal femur. Above these limits, a cyst was prone to fracture. Mechanically, a cyst was considered healed when the index was less than 3, with a cortical width more than 2 mm.

Cyst diameter method of Ahn and Park - A cyst occupying more than 85% of the bone diameter is at high risk of fracture.(11)

4. Joint incongruity

5. AVN

6. Pathological fracture

Radiographic differences between cystic lesions

	SBC	ABC	FIBROUS DYSPLASIA
Lucency	central	eccentric	central
Location	abuts physis	metaphyseal	metadiaphyseal
Appearance	uniloculated	multiloculated	ground glass
Width	same as physis	expansile	expansile

Cortex	thinned unless frac destroyed	thinned	
Calcification	rare	present	present
Cartilage	nil produced		
maturity	moves away	no change	no change
from physis			

B. CT –It is most helpful in evaluating the extent of pelvic cysts but can be used for evaluation of cysts in any location. When the cyst is found in the midshaft of a bone or in an unusual location, CT may be useful in determining the extent of the defect. If the CT scan is utilized to evaluate these cysts, Hounsfield units should always be included on the films to confirm the existence of fluid, since lipomas of bone also occur especially in the calcaneum, vertebral bodies and the proximal femur (the second most common location for cysts) have a similar plain film and CT scan appearance. Fluid within cystic defects is typically less than 20 Hounsfield units (2-18 U). Fat is less than 0 Hounsfield unit (0 to -200), and is helpful for differentiation.

Rising bubble sign-This sign indicates presence of a hollow lesion such as simple bone cyst in which a tiny gas bubble is formed at the time of pathological fracture and rises to the most non-dependent margin of the bone lesion. The bubble of gas can arise by one of two mechanisms: rapid tissue compression and decompression during trauma (a vacuum phenomenon) or by the establishment of communication

with the atmosphere (as seen in open fractures or instrumentation).It is seen in acute setting of a pathological fracture in simple bone cyst.(39)

C. MRI -Typical bone cyst will show a low [T1] and high [T2] signal on MRI. It is more easier to detect intracystic fluid or a single gas bubble than on CT. MRI is also used to exclude the presence of intralesional soft tissue or solid matrix. However, these findings do not clearly differentiate the aneurysmal bone cyst from the simple cyst in the child under age of eight years.(39)

D.Cystogram- Injecting a radio-opaque dye in the cystic defect as an additional diagnostic aid, stating that if this material fills the cavity, the diagnosis of a cyst can be safely made. A cyst that does not fill with dye implies a solid tumor. Limited recent reports also show that injection of fluid into the closed medullary cavity of the bone increases the intraluminal pressure fourfold. There has been one case of fatal embolism during injection techniques into the humeral canal for the treatment of a cyst. Inactive cysts occasionally have well-formed septations that prevent complete filling of the defect (especially if a fracture has previously occurred), thereby giving rise to a confusing picture when xray shows a simple bone cyst.(8)

DIFFERENTIAL DIAGNOSIS

Some lesions which should be considered in differential diagnosis are :- (32,38)

1.Fibrousdysplasia-Lesion in the metadiaphyseal region of the bone with well defined zone of rarefaction which is surrounded by a narrow rim of sclerotic bone. Sometimes, the tumor produces a large expansile mass that bulges in the soft tissue ex. Base of skull and maxilla .The tumor may have large amounts of cartilage which appears as dot or ring like calcification on radiographs.Sometimes, they have superimposed aneuysmal bone cyst.

2.Nonosteogenic fibroma-These are metaphyseal defects in the first and second decade of life, mostly in the distal femur. On radiographs, it usually present as lesions located eccentrically in long tubular bones with bulging of the cortical outline which appear to migrate towards the centre of the boneas the physis grows away from it. On histopathogenesis, they show a characteristic spindle cell proliferation with a loose storiform arrangement of cells. Usually no treatment is needed.

3.Giant cell tumor-These are epiphyseal tumors that usually arise in the second and third decade of life. Radiographs show expanding zone of radiolucency in the end of long bone of an adult. It usually extends to the articular cartilage,but there may be thin zone of normal bone intervening between the two. It frequently destroys the cortex extending to the soft tissues. On histopathogenesis, the characteristic cells are spindle shaped cells .Giant cells containing 40-60 nuclei are scattered through the lesion. Treatment usually consist of extended curettage and providing structural stabilisation to the bone.

4.Osteitisfibrosacystica- It is usually produced due to hyperparathyroidism. The lesion is filled with rich fibroblastic connective tissue rich in osteoclastic like giant cells resulting in resorption of bones resulting in weakening of the bones as their calcified supporting structures are replaced with fibrous tissue (peritrabecular fibrosis) and the formation of cyst-like brown tumors in and around the bone. Radiographs show thin bones which are bowed or fractured. Blood tests will show increased serum calcium and parathyroid hormone. Treatment is usually directed towards parathyroidectomy.

5.Enchondroma-It is a benign tumor of mature hyaline cartilage, usually located centrally in the metaphyseal bone. The incidence is evenly distributed in all the decades of life. Majority are located in the phalanges of the hand and feet. On radiographs, they produce a localised area of rarefaction. Long bone enchondromas are usually associated with mineralisation which is described as ring like or popcorn like. There is scalloping of the endosteal part of the cortex.Histopathogenesis shows presence of cartilage nodules with intervening bone trabeculae which lies in the marrow and does not involve the medullary bone. Usually no treatment is required or simple curettage is sufficient.

6.Neurofibroma-Neurofibromatosis is usually associated with various skeletal changes caused by contiguous neurogenic tumors like scoliosis, defects in the posterior orbital wall, congenital bowing and pseudoarthrosis.

7.Lipoma-Intraosseouslipomas are very rare and can involve any part of the skeleton with proximal femur the most common location. On radiographs,it presents as a well circumscribed area of radiolucency with central area of sclerosis. On MRI,

the fatty nature of the lesion becomes apparent. It may undergo malignant transformation.

MANAGEMENT

Since the first description of simple bone cyst in 1910, many methods of treatment have been tried with varying degrees of success.(2)

Initially Brunschwig et al in 1930 had described treatment of simple bone cyst with expectant management or in cases where there was pathological fracture, patients were usually treated with curettage of the lesion and swabbing out the lesion with antiseptic solution such as iodine.(13).

Aldredge et al in 1942 and Garceau et al in 1954 described treatment of simple bone cyst with various methods such as irradiation, curettage, curettage and bone grafting and various combinations. They found 100% recurrence with irradiation only. They found 33% recurrence rate using curettage alone. Their best results were with curettage and using bone chips. Aldredge et al used chemical cauterisation such as Zinc chloride and carbolic acid. An important finding in their study was that better results were obtained when cysts were treated in the latent phase.(32,34)

Neer et al in 1966 showed that operative treatment showed better rates of healing than non-operative treatment. They found immobilisation and restriction of activities did not help in treatment and 90% of the patients came back for surgery. In cases of humeral bone cyst, he reported that recurrences were twice more common in the age group of less than 10 years. Their treatment consisted of using

curettage,cauterization using zinc chloride, phenol and bone grafting. In their series,curettage and bone grafting without cauterisation showed 39% recurrence compared to curettage and bone grafting with cauterisation with a recurrence rate of 30%. They showed 31% recurrence using allograft compared to 23% when autogenous graft is used. (12)

Fahey et al in 1968 described subtotal resection of the cyst with bone grafting in which the involved area of the bone is exposed along with the cyst and transverse cuts are made on the bone adjacent to the proximal and distal end of the cyst upto $\frac{3}{4}$ th of the diameter of the width of the bone,curretting the cyst and filling it with bone graft. Similar study was done without bone grafts showing comparable healing rates.(9,16).

There were many complications associated with currettage and bone grafting such

- high recurrence rates (30-40%)
- pathological fracture
- limb length discrepancy (14%)
- premature epiphyseal closure(2-4%)
- infection(1%)(4,14)

This resulted in advancement of procedures with minimal surgical interventions like steroid injections, injection of demineralised bone matrix and autologous bone marrow or combination of these procedures.

Scaglietti et al in 1979 described the use of methylprednisolone in the treatment of simple bone cyst showing a recurrence rate of 10%. In another series, the injection was repeated in 76% of the patients with an average of three or four times with a maximum of nine injections. In their procedure, they injected 40-200mg of methylprednisolone in the cystic cavity. In the course of treatment, 2% of patients developed pathological fractures.(17,18).

Komiya et al in 1991 described a procedure drilling multiple puncture drill holes in the cyst (trepanation) after aspiration of the cyst fluid through a trocar. This method is effective because first, by removing cyst fluid, the internal pressure of the cyst becomes depressed, and the venous flow around the cyst wall is improved. Removing cyst fluid is also useful for removal of bone resorptive factors and, therefore, for inhibiting a progressive osteolysis. Second, making multiple drill holes through the cortical bone of the cyst wall stimulates the periosteum to induce bone formation and makes cyst fluid escape through the drilled holes. Third, making multiple drill holes through the medullary bone of the cyst wall improves the intramedullary venous flow. When a pathologic fracture occurs, cyst fluid escapes from the cyst and the periosteum is stimulated to accelerate bone formation. There was a recurrence rate of 27% using this procedure.(20)

Lokiec et al in 1996 reported 100% results with autologous bone marrow injection in the treatment of simple bone cyst using a trocar to disrupt the lining membrane, aspirating the cyst fluid and injecting bone marrow aspirate through the same trocar. A maximum of 25ml of aspirate was introduced in the cyst cavity.(19)

Docquier et al in 2003 did a similar study using autologous bone marrow with 12% recurrence rates.(4) Wright et al did a randomised control study comparing the healing rates in patients who were given either bone marrow injection or steroids as

a part of the trial. All the patients were a maximum of three injections. They showed that the healing rate was better in the steroid group compared to the bone marrow injection group and the risk of fracture was lower in the steroid group.(41)

As both steroids and bone marrow procedures showed comparable rates of recurrence of 10-15%, demineralised bone matrix was used more and more alone or in combination with bone marrow injections. Killian et al in 1998 used DBM for the treatment of simple bone cyst in which they obtained 22% recurrence.(42) Rourgraff et al in 2002 and Kanellopoulos et al in 2005 used DBM with bone marrow and showed a recurrence rate of 20-25%.Demineralized bone matrix is commercially available as an injectable substrate. It is prepared from freeze-dried allograft with a method that may preserve some of its osteoinductive and osteoconductive properties, and then it is mixed into a glycerine carrier, giving the material a gel-like consistency. This substance (Grafton; Musculoskeletal Transplant Foundation, Holmdel, New Jersey) is commercially available pre-packaged in syringes and can be injected through large-gauge needles. (6,8)

Peltier et al have operated on simple bone cyst with curettage and packing the cyst area with plaster of Paris pellets showing 90% success rates.(21)

The predictors of failure of treatment were described by Sung et al in 2008, which were:-

1. Younger age
2. Treatment with steroids

Gender, initial fracture and location of the cyst are not indicators of failure.(30)

Newer techniques such as bone scaffolds are increasingly being used over the last 10 years. ex. Tricalcium phosphate, calcium sulphate and high porosity calcium hydroxyapatite. Dormans et al in 2005 used the technique of percutaneous intramedullary decompression, curettage and grafting using MGCS pellets showed a healing rate of 91.7%. Inoue et al in 1993 have described the use of high porosity hydroxyapatite in filling the cavity of the cyst after curettage. It is composed of calcium hydroxyapatite $[\text{Ca}_{10}(\text{PO}_4)_6(\text{OH})_2]$. The HA is composed of 70% of highly porous connecting air cells of 90 μm in diameter and sintered at 900° . The authors have found a healing of 78% using high porosity HA. They are used for their osteoinductive and osteoconductive properties accelerating the rate of healing after decompression of the cyst by various methods. The advantage over traditional bone grafting methods are decreasing the donor site morbidity in children, decreased chance of infection and more quantity obtained to fill the cyst. (22,23,24)

The recurrence rates with the various techniques were:-

- Expectant management- 80-100% (13)
- Irradiation - 90-100% (13, 32)
- Curettage alone- 50-60% (12, 32, 34)
- Curettage with bone grafting- 30-40% (12, 27)
- Curettage, bone grafting and chemical cauterisation- 30-35% (12)
- Curettage and cortical\ cancellous allograft- 12-45% (33)

- Subtotal resection with or without bone grafting- 5-10% (35)
- Trepanation- 20-25% (5)
- Steroids \ autogenous bone marrow - 10-15% (18, 41)
- Demineralised bone matrix – 20-25% (6, 8 ,30)
- POP pellets- 12% (21)
- Bone ceramics such as calcium sulphate or hydroxyapatite- 10% (22, 24)

In patients presenting with pathological fracture, they were usually treated with immobilisation casts till bone healing was visualised on radiographs. Bumci et al have used Kuntscher nail in displaced fractures with or without cerclage wires achieving comparable results.(10)

As the recurrence rates were high and unpredictable in the above mentioned techniques,management has evolved towards flexible intramedullary nail as the method of treatment of simple bone cyst. One of the main advantages of using an intramedullary nail was the absence of postop immobilisation. A review of the literature showed that approximately 50 % of patients with a bone cyst have a complete pathological fracture and another 25 % have infraction of a thinned cortex at the time of presentation. Although local methods such as steroid injections, bone-marrow injections, and decompression produce consolidation in most patients, they offer no immediate mechanical stability to the weakened bone. Nailing has the benefits of decompression and providing early stability to the bone, which permits

early mobilization, thus obviating the need for a plaster cast and decreasing the prevalence of the most common complication; a pathological fracture. (27,43,46)

Catier et al in 1981 first described the use of Ender nails in fixation of pathological fractures due to simple bone cyst in the femur.(28) Santori et al used flexible IM nailing in 1986 for pathological fracture in the proximal humerus and femur achieving 100% healing rates. Roposch et al in 2000 operated in 32 children with flexible intramedullary nailing, out of which 30 presented with pathological fracture. Healing in these patients were assessed with Capanna's grading and 30 of the 32 patients had shown grade 1 and 2 healing. They had a recurrence rate of 6%. They had another 6% recurrence after removal of the nail. The other complications they encountered were change of nails in 28% of patients because of shortening in the growing bone and varus deformity of the proximal femur in 15% of the patients. Sanctis et al in 2006 operated on 47 patients with simple bone cyst with no recurrences. All the lesions had healed, 60% completely and 40% with residual radiolucency according to Capanna's grading. In two patients where the cyst was close to the physis, there was evidence of limb length discrepancy of 1cm. Pogorelić et al did not find any recurrence in their 18 patients in a followup of 1.5-5 years. Their only complication was irritation at the nail entry site. Kanellopoulos et al in 2007 used flexible intramedullary nailing in simple bone cysts with DBM and bone marrow to fill the cystic cavity in 9 patients. They introduced the DBM into the cyst cavity after calculating the cyst ratio. In their study, they reported no recurrence in a 5 year followup with the only complication, being, irritation at the nail entry site. The advantage of using flexible intramedullary nail is that it provides continuous decompression of the cyst and acts as an internal splint. (25,26,27,43,44)

As shown in the above studies, flexible intramedullary nailing in simple bone cyst are associated rarely with complications. The recurrence reported in one study was 6%,rest of the studies did not show any recurrence. The major complication reported in the above studies was varus deformity at the fracture site, change of nail due to shortening only in one study and irritation at the nail entry site.

MATERIALS AND METHODS

All cases of children admitted with a diagnosis of simple bone cyst between January 2004 to December 2009 were identified from the operation register, biopsy reports and in-patient records. Discharge summaries were searched from the clinical workstation, a hospital information archival and retrieval software. Radiographs of these patients were obtained from PACS.

38 children, in the Paediatric Orthopaedic unit, who were under the age of 18 years, were identified to have a diagnosis of simple bone cyst of femur, humerus and radius. 28 had undergone flexible intramedullary nailing, 26 of these had cyst filled with commercially available bone substitutes (hydroxyapatite or calcium sulphate). The other 10 had undergone treatment with alternative techniques and do not form part of this study.

Postoperative follow up was available on 27 patients and one was lost to follow up after the surgery. The mean duration of follow up was 16 months and ranged from 3 months to three and a half years. The patient who has no follow up was excluded from this study.

The pre-operative data collected from these patients included site, age, gender, location of the cyst, activity of the cyst, presence or absence of pathological fracture, use of bone substitutes and any prior surgeries. The postoperative data collected from these patients were duration of followup, radiographic signs of healing and presence or absence of postop complications.

The choice of treatment was made by the paediatric orthopaedic surgeon. The size of the titanium nails to be used was determined by measuring the smallest diaphyseal

internal diameter of the bone in both the antero-posterior and lateral radiographs and this was divided by 2 and 1 mm subtracted to arrive at the nail diameter to be used.(37)

OPERATIVE TECHNIQUE

Proximal femur

The patient was placed in supine position on the standard operating table. After the extremity has been surgically prepped and draped, the proximal femur cyst was located under image intensifier and aspirated. A clear fluid aspiration confirmed the diagnosis of simple bone cyst. In case of a pathological fracture or a blood filled cyst the cyst was opened for grafting. In other cases an opening was made and biopsy obtained to rule out other cystic lesions such as aneurysmal bone cyst and fibrous dysplasia.

When opening the cyst the incision was made on the lateral aspect of the thigh, dividing the fascia reaching the anterolateral cortical wall of the cyst. Now, a cortical window is made on the most prominent part of the cyst wall draining the cyst fluid and a thorough curetting the membranous lining of the cyst wall is carried out. Distally symmetrical skin incisions are made on the medial and lateral side. The distal skin mark was the upper pole of the patella and progressed 2-3cm proximally. The skin and the fascia were incised together. Blunt dissection was continued through the muscle to the bone. The entrance points for nail should be outside the joint capsule and away from the edge of the physis. The entry sites were perforated by an awl in the most proximal end of the incision 2-3cm proximal to the physis. The

awl was initially placed at 90 degrees to the cortex to keep it from slipping off. Once the awl was firmly seated on the cortex, it was reduced to an angle of 45 degrees to the shaft axis and the perforation of the bone was continued at an upward angle. Carefully the pre-bent nail with a curve at the tip was inserted into the medullary canal by hand or using the T – handle inserter. Following its insertion, the position of the nail was confirmed with the image intensifier. The curve of the tip was accentuated to facilitate its bouncing off the opposite cortex. Carefully the first nail was advanced up to the region of pathological fracture zone. Following this the second nail was inserted from the medial side and advanced to the cyst. At the cyst the nail was rotated three hundred sixty degrees and advanced in increments to provide an opening of all septae usually present in children with previous fractures. Both nails were advanced into the proximal fragment crossing the proximal cyst wall. Once the nails cross the cavity, it is packed manually with bone substitutes. The medial nail was directed to the femoral neck and the lateral nail toward the greater trochanter and an attempt is made to have a purchase in the physis. This prevents backing out of the nail. Just prior to advancing to their final position, the nails were cut and terminal part bent to lie against the bone, leaving enough length to manipulate and advance them to their final position. Once both the nails have entered the proximal part they were then tapped to their final position. Once the nail tips were in their final position, the end of each nail was cut, leaving 1-2cm protruding from the cortex. The cyst size and adequacy of curettage is measured by a cystogram and then the cyst is packed with bone substitute. The wounds are closed with drains.

Postoperatively, the child is ambulated on the second post operative day partial /non weight bearing with crutches for 4-6 weeks. The x-rays were taken in the outpatient

clinic at six weeks to three months. If there are sufficient signs of healing, full weight bearing is started. During follow up we recorded the range of motion of hip and knee, radiographic healing, limb length discrepancy, the condition of wound and skin, other symptoms and other complications.

Radiographic healing was measured according to Capanna's grading.

2. Proximal humerus - Patient lies supine on the radiolucent table. After localising the cyst under image intensifier, the cyst is opened through a mini incision (2.5 to 5 cm) in the deltopectoral groove. The most affected cortex is opened, the cyst fluid is drained and the cyst lining is aggressively curetted and cyst volume and quality of curetting assessed by cystogram. A single humeral nail is used in cysts with no fracture or sufficient structural integrity. Distally, one incision is made on the lateral aspect of the distal humerus. The entrance point for nail should be outside the joint capsule and away from the edge of the physis. The entry site was perforated by an awl in the most proximal end of the incision. The awl was initially placed 90 degrees to the cortex to keep it from slipping off. Once the awl was firmly seated on the cortex, it was reduced to an angle of 45 degrees to the shaft axis and the perforation of the bone was continued at an upward angle. Carefully the nail was inserted into the medullary canal by hand. Following its insertion, the position of the nail was confirmed with the image intensifier. The curve of the tip was accentuated to facilitate its bouncing off the opposite cortex. Carefully the nail was advanced up to the region of pathological zone. The nail is advanced into the proximal fragment crossing the proximal cyst wall as described under the femur technique. Once the nail crosses the cavity, it is packed manually with bone substitutes.

Postoperatively, the patient was kept in a broad arm sling for a period of 2-3 weeks.

Xrays were taken serially at 6 weeks, 3 months, 1 year for followup and periodically after that for unresolved cysts to assess the healing according to Capanna's grading.

ASSESSMENT OF OUTCOME

Radiological assessment

Capanna's grading(46):-

Grade 1-healed when cyst is completely filled with bone and cortical margins are thickened.

Grade 2- healed with residual radiolucency when most of the cyst is filled with bone and cortical margins are thickened, but there were small residual areas of radiolucency.

Grade 3- recurrence, when cyst had healed initially and had filled with bone but large areas of radiolucency and cortical thinning subsequently developed.

Grade 4 –no response when there is no evidence of any effect of treatment.

Radiographic assessment of healing was taken at the minimum duration when the cyst showed the best grade of healing and the minimum period for evaluation was at 3 months. (26,44)

RESULTS

Flexible intramedullary nailing was done in (21 children in the proximal humerus, 6 in the proximal femur) for a confirmed diagnosis of simple bone cyst in children in the paediatric unit in Christian Medical College and Hospital, Vellore.

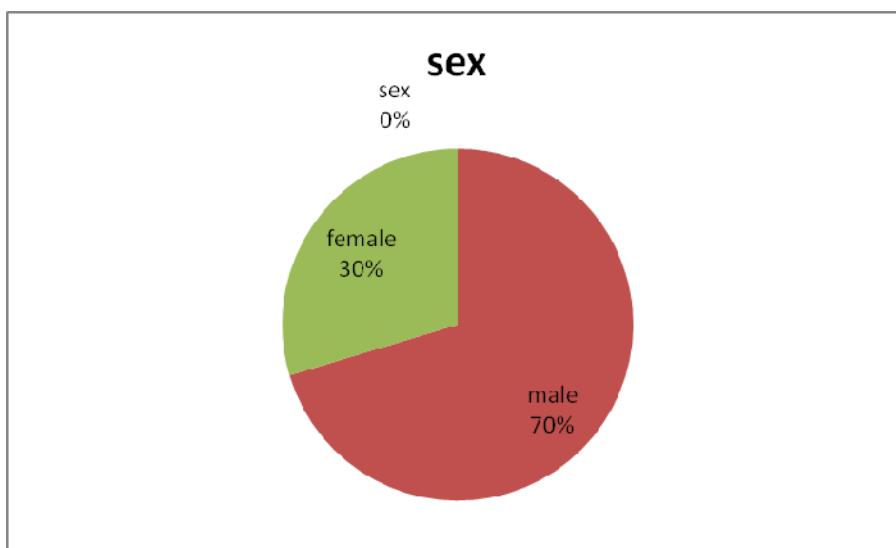
27 patients with followup were included in the study. There were nineteen males and eight females. The mean age of the patients is 10.3 years ranging from 3 to 18 years. The mean duration of postoperative stay in the hospital was 4.9 days.

All were symptomatic with symptoms ranging from ache, swelling, decreased range of motion and inability to walk (for femur) or use upper limb.

1. Sex ratio

Male=19

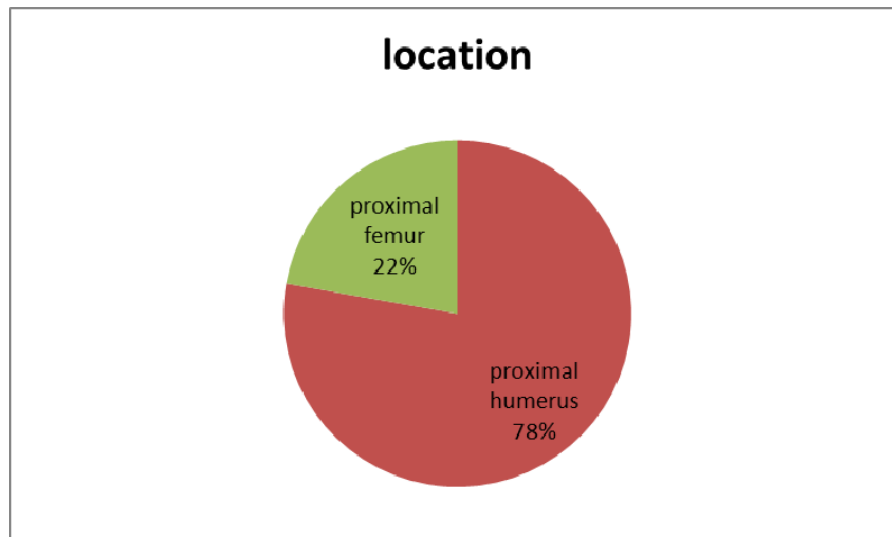
Female=8



2. The location of cysts

Proximal humerus=21

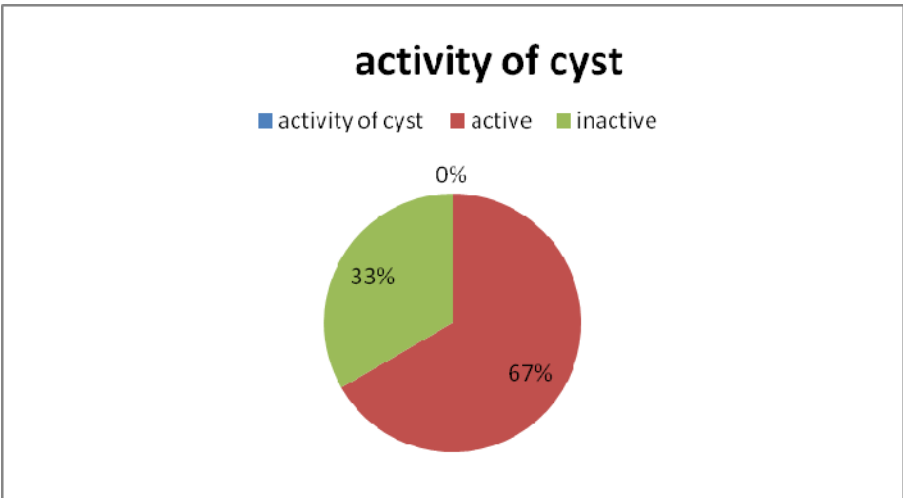
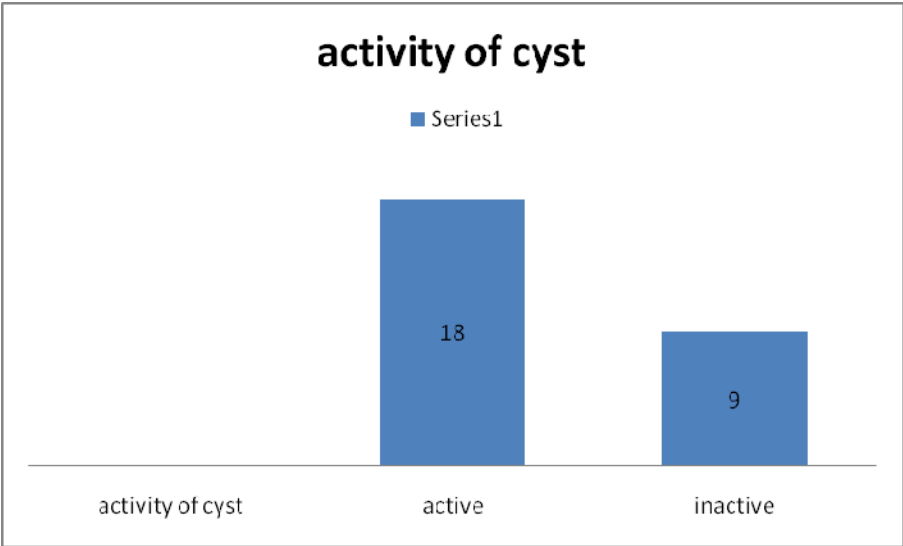
Proximal femur=6



3. Activity of the cyst

Active = 18

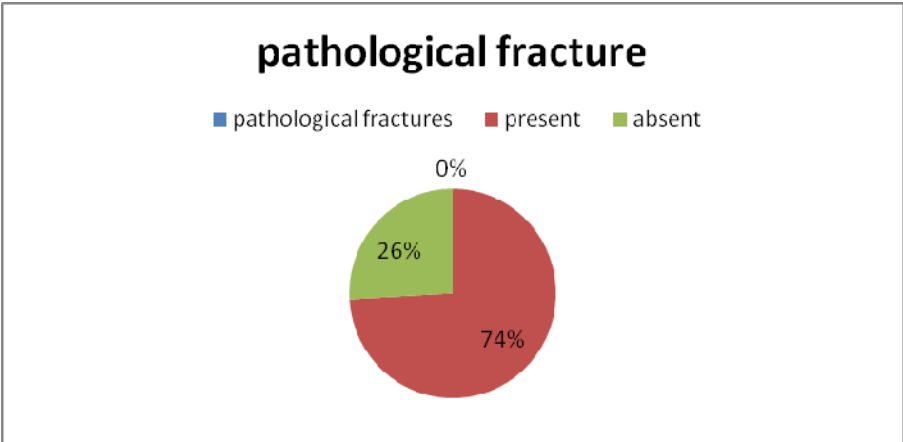
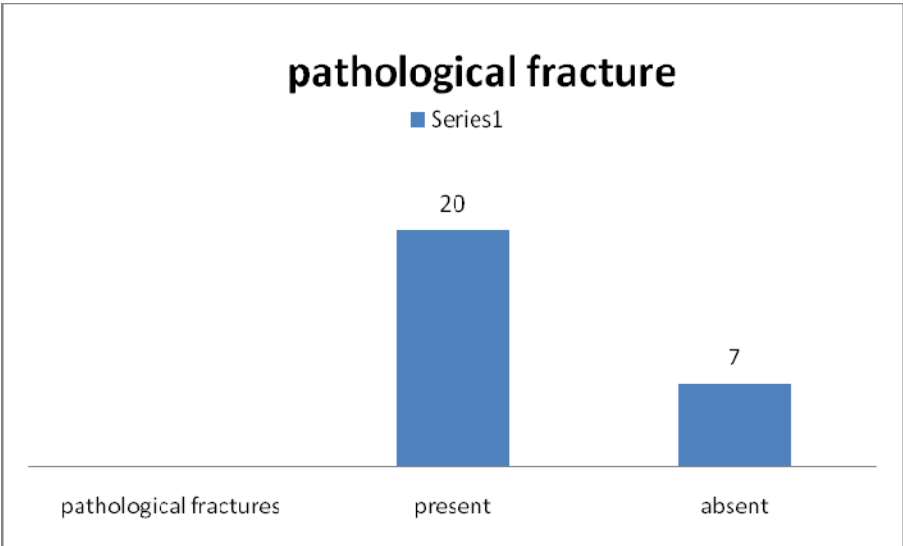
Inactive =9



4. Presence of pathological fracture

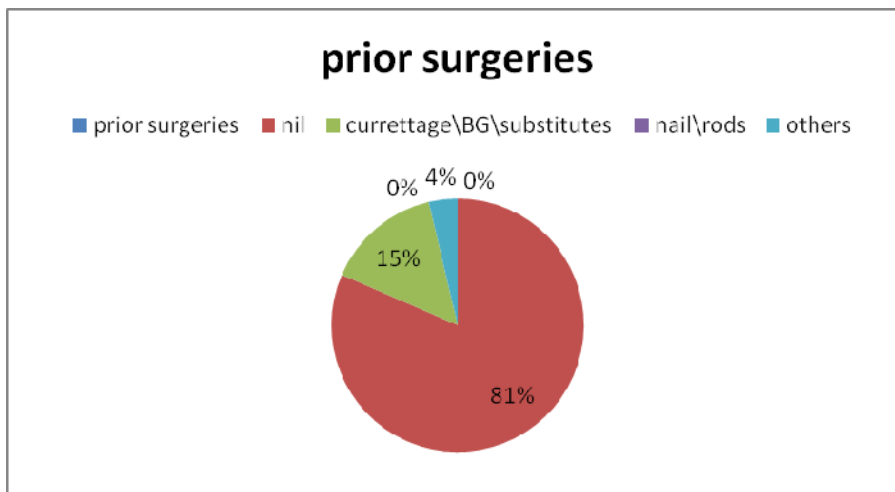
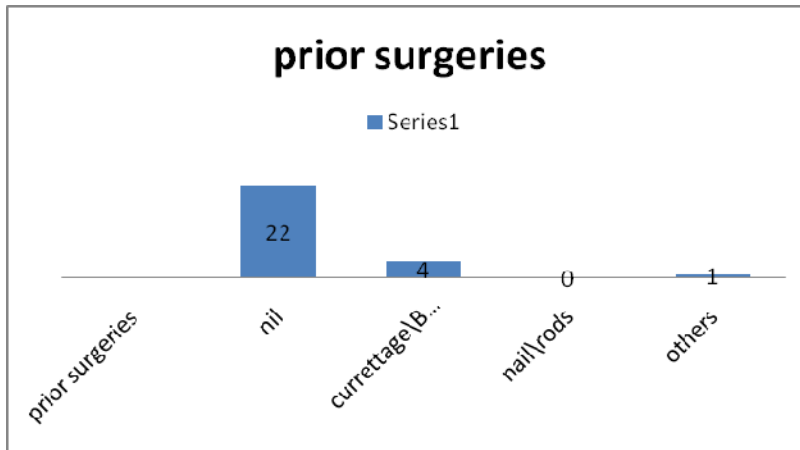
Present =20

Absent = 7



5. Prior surgeries

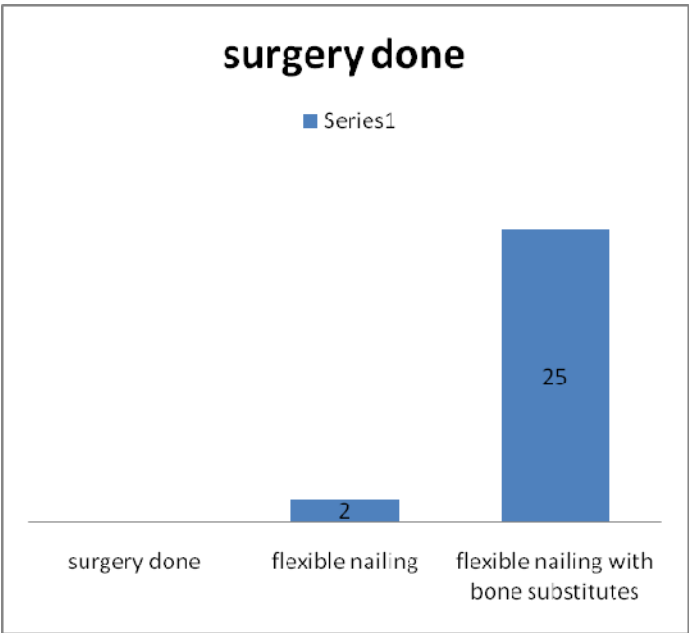
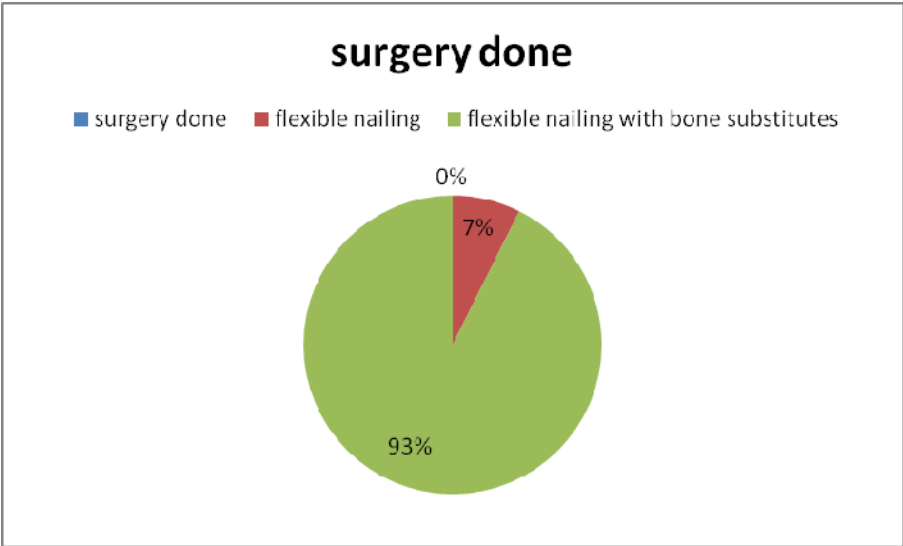
Nil	22
Currettage\BG\substitutes	4
Nail\rods	0
Others	1



6. Surgery done

Flexible nailing with bone substitutes=25

Flexible nailing only =2



7. Duration of followup

3months =4

6months =4

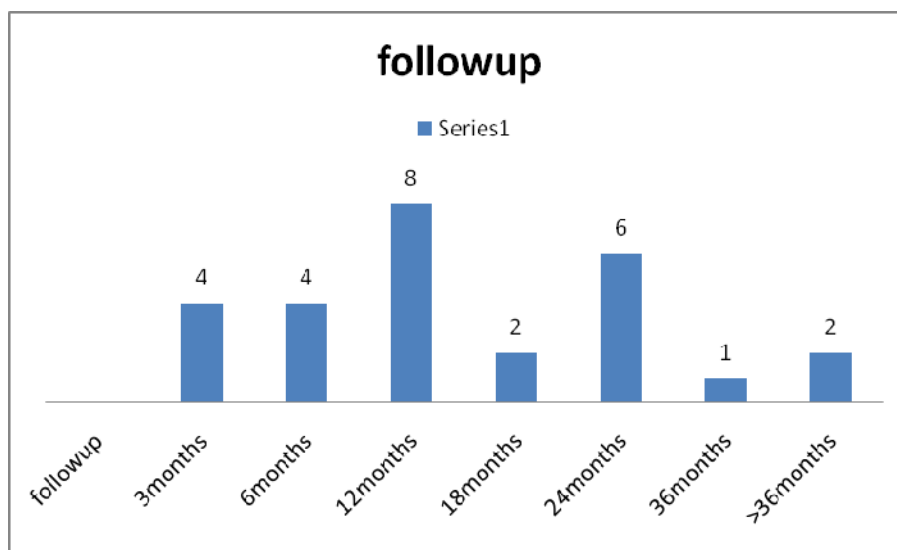
12months =8

18months =2

24months = 6

36months= 1

>36months= 2



8. Assessment of healing

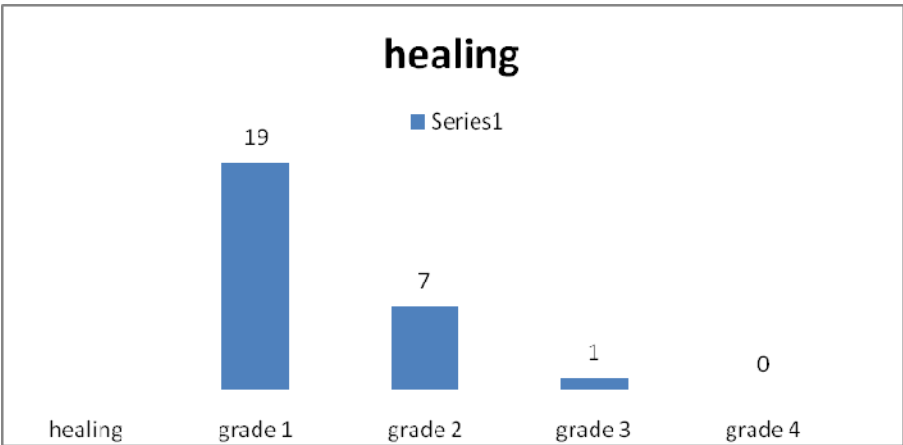
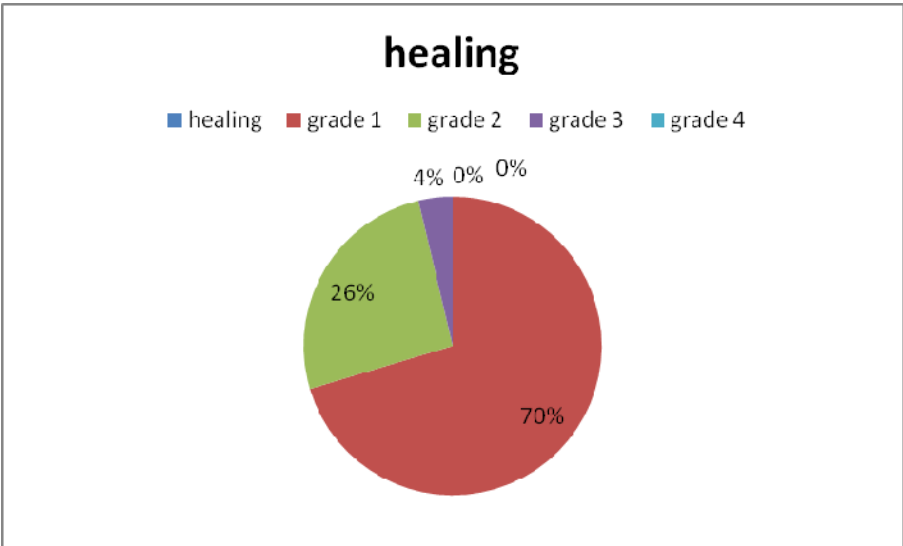
Healing by Capanna's grading

grade 1 =19

grade 2 =7

grade 3 =1

grade 4 =0



9.Complications

There were no complications in 23 children Four children had 5 complications requiring repeat surgery.

Nil= 23

Recurrence = 3

Angular deformities =0

Physeal damage =0

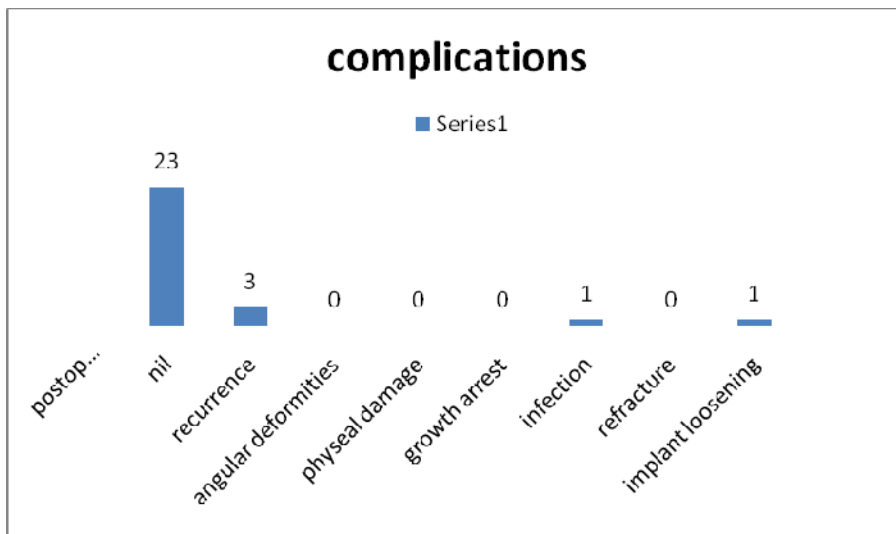
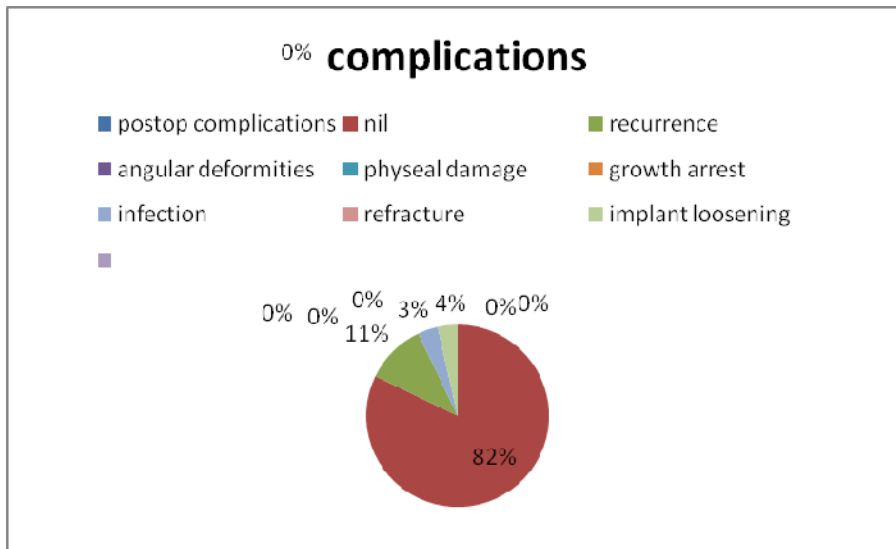
Growth arrest =0

Infection =1

Re-fracture =0

Implant loosening =1

One had infection of the nail which was treated with implant removal. The cyst recurred after one year and a second nailing procedure was done for the recurrence. Another child had a recurrence of the cyst in the femur and this was treated with rush rod and bone substitute filling of the cyst. The third child developed a second cyst occurred at a geographically different level distal to the previous site and this was treated with a bigger size nail and grafting. The fourth patient had implant loosening which was removed as the cyst had healed.



DISCUSSION

Many treatment methods exist in the literature showing that as of now there is no widely accepted effective treatment for simple bone cyst .(30) Flexible intramedullary nailing to drain and heal the cyst has been described three decades ago but has gained acceptance only recently. However the time to heal for the cyst by this technique is still averages about 5 months. Since these lesions limit the patient activity on account of a fear of pathological fractures we have added bone substitutes as they do not increase the morbidity or risk but increase the strength of the affected area during the phase of healing. This study looks at the outcome of this previously undescribed combination of interventions in simple bone cyst in children. Postop range of motion was almost fully obtained during the time of follow up of these patients. In our study we have found the sex ratios showing preponderance of male to be similar to the sex ratio in literature.(4) The mean age of the children in our study was 10.3 years which was similar to that described in the literature.(27,43)

The site of the lesion is predominantly proximal humerus in most studies which is similar to the findings of our study as 78% of our children had a cyst in the proximal humerus.(12,32)

2\3rd (66.6%) of the cysts in our study were present in the active phase which were more than studies done by Roposch et al and Sanctis et al.(27,43) This is probably because this study only involves children. As many as 74% had an event of fracture before presenting here which is again comparable to the findings in the literature.

Recurrence is the hall mark of simple bone cyst and out of 27 patients in our study, 4(15%) had a prior surgical intervention such as curettage and bone grafting\bone substitutes. One patient had an external fixator in the arm after been diagnosed with

pathological fracture. In studies done by Roposch et al and Sanctis et al no patients had previous surgery. Those who presented with pathological fracture were managed conservatively with cast immobilization before being included in their study.

In our study 25 patients(93%) had undergone flexible intramedullary nailing and bone substitutes to fill the cystic cavity. In literature, the only similar study is by Kanellopoulos et al. They however used demineralised bone matrix with added bone marrow in addition to putting the flexible nail. Roposch et al and Sanctis et al did not use any bone substitutes to fill the cystic cavity.

In our study 19 patients (70%) showed Capanna's grade 1 healing, 7 patients (26%) showed grade 2 healing and 1 patient (1%) showed a grade 3 outcome on radiographs. The grade 3 child was characterized as a recurrence and underwent second surgery. These results are comparable to those published in the literature for flexible intramedullary nail in literature. Roposch et al in a series of 32 patients have shown 6% recurrence rates. We had 11% recurrence, 4% infection and 4% implant loosening.

Many treatment strategies have been tried for simple bone cyst till now which includes curettage and bone grafting, subtotal resection, trepanation and microdrilling, use of autogenous bone marrow demineralised bone matrix and use of artificial bone substitutes like calcium phosphate and hydroxyapatite and use of steroids with overall recurrence rates on 15-20%. Each of these modes of treatment are associated with various complications like physeal damage, growth arrest and infections.

One important shortcoming in the various methods used to treat simple bone cyst in literature was the inability to describe methods to provide internal splinting as a part of the treatment. This is very important as it limits the activities of daily living in patients if the cyst occurs in the weight bearing bones. This is the distinct advantage of using flexible intramedullary nail in the treatment as it acts an internal splint for weight bearing as well as providing continuous decompression.

One of the presently popular techniques at present of using repeated steroid injections is difficult in the Indian population as patients are unable to travel long distances for repeated followup, they need support to avoid pathological fractures or limit their activities and multiple anesthesia make the procedure more expensive. Steroid injections are also associated with various complications such as avascular necrosis, pathological fracture during the course of treatment and LLD. In literature, a recurrence rate of 12-15% is documented and the healing is unpredictable. (46)

In cases of pathological fractures, the affected patients are either treated with cast or immobilisation. K nails have been described for treatment of pathological fractures of the femur. The use of flexible intramedullary nail for the treatment of simple bone cyst provides fixation by internal splinting and has been associated with excellent results. In 2006 Sanctis et al in 47 and Kanellopoulos et al in 9 patients obtained 100% results with the only complication being irritation at the nail entry site. Roposch et al have shown a small recurrence rate.

In this study, the recurrence rate is 11% with a low complication rate. This makes it a suitable form of treatment for this condition. From this study since most (except 2)

patients also received some form of bone substitutes it is difficult to compare and say if it was really necessary.

On the basis of these results, we posit flexible Intramedullary nailing with and without the addition of bone substitutes an efficient and safe procedure for the treatment of simple bone cyst.

CONCLUSION

This study validates the result of 27 patients treated with flexible intramedullary nail with or without bone substitutes. We conclude this treatment regime is a safe and effective way to treat simple bone cyst in children.

SCOPE OF THE STUDY

This is a retrospective review, thus it has its limitations. We were unable to completely assess the healing time because of presence of the scaffolds. A randomized controlled trial comparing this technique with other techniques of treatment in our country and looking at the cost effectiveness of various techniques might give us a better insight into which is the best method of treatment in simple bone cyst in India at present.

BIBLIOGRAPHY

1. **Broder, H** Possible Precursor of Unicameral bone Cysts *JBJS A* 1968;50:503-507
2. **Bloodgood, JC** Benign bone cysts, osteitisfibrosa, giant cell sarcoma and bone aneurysm of long pipe bone. *Annsur* 1910;52:145-89
3. **Jaffe, HL and Lichtenstein, L** Solitary unicameral bone cyst with emphasis on the roentgen picture: the pathological appearance and pathogenesis. *Arch Sur* 1942;44:1004-25.
4. **Docquier,P and Delloye,C**Treatment of Simple Bone Cysts With Aspiration and a Single Bone Marrow Injection *JPO A* 2003;23:766–773
5. **Komiya, S ; Inamitan, K ; Asaguri, Y; Ashimoto, S ; Orimatsu, M and Inoue, A** Simple Bone Cyst Treatment by Trepanation and Studies on Bone Resorptive Factors in Cyst Fluid With a Theory of Its Pathogenesis *CORR February. 1993* 204-211
6. **Kanellopoulos,A ; Yiannakopoulos, CKand Soucacos,P** Percutaneous Reaming of Simple Bone Cysts in Children Followed by Injection of Demineralized Bone Matrix and Autologous Bone Marrow *JPO A* 2005;25:671–765
7. **Cohen, J** Simple Bone Cysts: Studies of Cyst Fluid in Six Cases with a Theory of Pathogenesis *JBJS A* 1960;42:609-616
8. **Rougraff, B and Kling,TT**Treatment of Active Unicameral Bone Cysts with Percutaneous Injection of Demineralized Bone Matrix and Autogenous Bone Marrow *JBJS A. 2002;84:921-929.*

9. **Fahey, J and O'Brien, E** Subtotal Resection and Grafting in Selected Cases of Solitary Unicameral Bone Cyst *JBJS A* 1973;55:59-68.
10. **Igor, B and Tomislav, V** Significance of Opening the Medullar Canal in Surgical Treatment of Simple Bone Cyst *JPO A* 2002
11. **Vasconcellos, D and Yandow, S** Cyst Index A Nonpredictor of Simple Bone Cyst Fracture *JPO A* 2007;27:307-310
12. **Neer, C ; Francis, K ; Marcove, R ; Terz, J and Peter, N** Treatment of unicameral bone cysts. A follow-up study of one hundred and seventy-five cases. *JBJS A*. 1966;48:731-45
13. **Brunschwig, A** Histology of solitary bone cyst of long duration *JBJS A* 1930;12:141-149
14. **Campanacci, M ; Campanna, R and Picci, P** Radiology xray of Unicameral and aneurysmal bone cysts. *CORR* 1986;204:25-36.
15. **Struhl, A ; Pritzker, H and Seimon, LP** Solitary (unicameral) bone cyst. The fallen fragment sign revisited. *Skelet Rad* 1989;18:261-265
16. **McKay, DW and Nason, SS** Treatment of unicameral bone cysts by subtotal resection without grafts *JBJS A* 1977;59:515-519
17. **Scaglietti, O; Marchetti, PG and Bartolozzi P.** The effects of methylprednisolone acetate in the treatment of bone cysts. Results of three years follow-up *JBJS B* 1979;61:200-4

18. **Scaglietti, O ; Marchetti, PG and Bartolozzi, P** Final results obtained in the treatment of bone cysts with methylprednisolone acetate (depo-medrol) and a discussion of results achieved in other bone lesions. *CORR.* 1982;165:33-42.

19. **Lokiec, F ; Ezra, E ; Khermosh, O and Weintraub, S.** Simple bone cysts treated by percutaneous autologous marrow grafting. *JBJS B* 1996;78:934-7

20. **Komiya, S ;Tsusuki, K and Mangham, D** Oxygen scavengers in simple bone cysts. *CORR.* 1994;308:199–206

21. **Peltier LF and Jones RH** Treatment of unicameral bone cysts by curettage and packing with plaster-of-Paris pellets. *JBJS A* 1978;60:820-822

22. **Dormans,JP ;Sankar, W ; Moroz,L and Erol,B** Percutaneous Intramedullary Decompression, Curettage, and Grafting With Medical-Grade Calcium Sulfate Pellets for Unicameral Bone Cysts in Children A New Minimally Invasive Technique *JPO A* 2005;25:804–811

23. **Wilkins, RM.** Unicameral bone cysts. *J Am AcadOrthopSurg* 2000;8:217-224

24. **Inoue, O ; Ibaraki, K and Shimabukuro, H** Packing with high-porosity hydroxyapatite cubes alone for the treatment of simple bone cyst. *CORR* 1993;293:287-292

25. **Santori,FS ;Ghera, S and Castelli,V**Possibilita` di guarigione di estese cisti Ossee giovanili trattate mediante iniezione di idrossidrossifosfato di calcio: interpretazione patogenetica. *IJOT.* 1986;12(4):429-436

26. **Pogorelić, Z ; Furlan, D ; Biočić, M ; Meštrović, J; Jurić, I and Todorčić, D**
Titanium Intramedullary Nailing for Treatment of Simple Bone Cysts of the Long
Bones in Children *SCM August 2010*
27. **Roposch, A ;Saraph, V and Linhart, WE** Flexible intramedullary nailing for the
treatment of unicameral bone cyst in long bones. *JBJS A2000;82-A:1447-1453*
28. **Catier, P ;Bracq, H and Canciani, JP** Indication inhabituelle de
l'enclouage de Ender: le kyste osseux fémoral de l'enfant. *Rev Chir
Orthop. 1981;67:147-149*
29. **Givon,U ;Sher-Lurie, N ; Schindler , A and Ganel, A**Titanium Elastic Nail—a
Useful Instrument for the Treatment of Simple Bone Cyst *JPO A 2004;24:317–318*
30. **Sung, A ; Anderson, M ; Zurakowski D; Hornice , F and Gebhardt, M**
Unicameral Bone Cyst A Retrospective Study of Three Surgical Treatments *CORR
(2008) 466:2519–2526*
31. **Cohen, J** Etiology of Simple Bone Cyst *JBJS A 1970;52:1493-1497*
32. **Garceau, GJ and Gregory, CF.** Solitary unicameral bone cyst. *JBJS A
. 1954;36:267–280*
33. **Spence, K ; Bright, C and Fitzgerald. P:** Solitary unicameral bone cyst:
Treatment with freeze-dried crushed cortical-bone allograft. *JBJS A 58A:636. 1976*
34. **Aldredge, R** LOCALIZED FIBROCYSTIC DISEASE OF BONE: Results of
Treatment in One Hundred and Fifty-Two Cases *JBJS A 1942;24:795-804*

- 35 **Spence, K ; Sell, K and Brown,R** Solitary Bone Cyst: Treatment with Freeze-Dried Cancellous Bone Allograft: A STUDY OF ONE HUNDRED SEVENTY-SEVEN CASES *JBJS A*. 1969;51:87-96
- 36.**Dormans, JPP**Pediatricorthopaedics Core knowledge in orthopaedics *Elvesier Mosby* 2005
- 37.**Kasser, J and Beaty, J** Femoral shaft fractures in children. eds. *Rockwood and Wilkin's . Lippincot Williams and Wilkins. Fractures in children . 6th edition* 2006
38. **Unni,K and Inwards, C**Dahlin's bone tumor *Williams and Wilkins. 6th edition*
- 39.**Jordanov, M** The "rising bubble" sign: a new aid in the diagnosis of unicameral bone cysts *Skelet Rad* (2009) 38:597– 600
40. **Schreuder, H; Conrad, E; Bruckner, J ; Howlett, A and Sorensen, L**Treatment of Simple Bone Cysts in Children with Curettage and Cryosurgery *JPO A* November/December 1997, pp 814-820
41. **Wright, J ;Yandow, S ; Donaldson, S and Marley, L** A Randomized Clinical Trial Comparing Intralesional Bone Marrow and Steroid Injections for Simple Bone Cysts *JBJS A* 2008;90:722-730.
42. **Killian, J; Wilkinson, L; White, S and Brassa,M** Treatment of Unicameral Bone Cyst with Demineralized Bone Matrix *JPO A*September/October 1998, pp 621-624
43. **Sanctis, NandAndreacchio,A** Elastic Stable Intramedullary Nailing Is the Best Treatment of Unicameral Bone Cysts of the Long Bones in Children? Prospective Long-term Follow-up Study *JPO A* 2006;26:520-52

44. **Kanellopoulos, A ;Mavrogenis, A ; Papagelopoulos, P and Soucacos, P**
Elastic intramedullary nailing and DBM-Bone marrow injection for the treatment of
simple bone cysts *WJSO* 2007
- 45**Barry, M and Paterson, J. M. H** Flexible intramedullary nails for fractures in
children *JBJS B SEPTEMBER* 2004
46. **Capanna, R; Dal Monte, A; Gitelis, S and Campanacci, M** :The natural history
of unicameral bone cyst after steroid injection. *CORR* 166: 204-211, 1986
47. **Ovadia,D ; Ezra, E ; Segev,E ; Hayek, S ; Keret, D ; Wientroub,S and
Lokiec,F** Epiphyseal Involvement of Simple Bone Cysts *JPO A* 23:222–229 © 2003

ANNEXURE

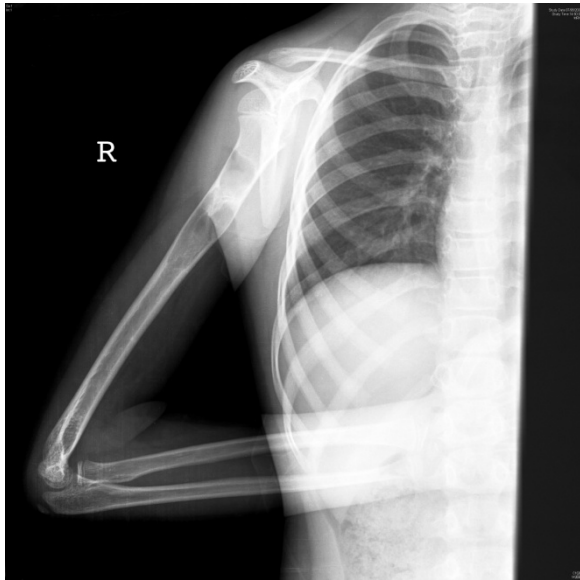


Figure1



Figure 2

Figure 1 & 2 showing pre-operative radiographs of patient 1 with lytic lesion in the proximal humerus which is epiphyseal in location and abutting the physis. There is expansion of the cortex. There is no pathological fracture.



Figure 3



Figure 4

Figure 3 & 4 of patient 1 showing intramedullary nailing for simple bone cyst in the proximal humerus 1 and 2 years post-operatively ,respectively, both of which show complete obliteration of the lesion and incorporation of the bone substitutes indicating grade 1 healing (Capanna's).



Postoperative photographs of patient 1 with healed scar on the right arm and full range of motion in both flexion and extension of the right arm



Figure 5



Figure 6

Figure 5 & 6 showing lytic lesion of the proximal humerus of patient 2 which is abutting the physis and associated with expansion of the cortex. There is pathological fracture of the lateral cortex.



Figure 7



Figure 8

Figure 7 & 8 showing flexible intramedullary nailing of simple bone cyst of proximal humerus of patient 2, 18 months postoperatively which has completely healed with grade 1 healing (Capanna's).



Post-operative photographs of patient 2 with healed scar on the right arm and full range of motion of the right arm in both flexion and extension.



Figure 9

Pre-operative radiograph of a lytic lesion of the proximal humerus of patient 3 which is abutting the physis and showing infraction of the cortex.



Figure 10



Figure 11

Figure 10 & 11 showing 18 months postoperative radiographs of patient 3 with reformation of the cyst just distal to the prior cyst .The first cyst has healed with residual radiolucency (Capanna's grade 2 healing). Redo nailing with bone substitutes was done for the new cyst in this patient.



Figure 12

Figure 12 showing 1 year postoperative radiograph after the second surgery of patient 3 which shows complete obliteration of the second cyst and incorporation of the graft (Capanna's grade 1 healing).

PROFORMA

NAME :

AGE:

SEX :

HOSPITAL NO:

DURATION OF FOLLOW UP:12WEEKS(3 MONTHS)

24 WEEKS(6MONTHS)

48WEEKS(12MONTHS)

72 WEEKS(18MONTHS)

98WEEKS(24MONTHS)

144WEEKS(36MONTHS)

MORE THAN 3 YRS

LOCATION:PROXIMAL HUMERUS

PROXIMAL FEMUR

SURGERY DONE : FLEXIBLE NAILING ALONE

FLEXIBLE NAILING AND BONE SUSBTITUTES

PRIOR SURGERIES: NIL

CURRETTAGE \BG\BONE SUBSTITUTES

RUSH RODS

FLEXIBLE NAILS

OTHERS

RADIOLOGICAL HEALING : CAPANNA'S STAGING

1=HEALED (COMPLETELY HEALED AND CORTICAL MARGINS HAVE THICKENED)

2=HEALED WITH RESIDUAL RADIOLUCENCY (MOST OF THE CYST HAVE HEALED BUT THERE ARE SOME AREAS OF RADIOLUCENCY)

3=RECURRENCE (INITIALLY HEALED BUT LATER THERE WERE LARGE AREAS OF RADIOLUCENCY WITH CORTICAL THINNING)

4=NO RESPONSE (NO EFFECT OF TREATMENT)

PATHOLOGICAL FRACTURE: ABSENT
 PRESENT

STATUS OF CYST ACTIVE
 INACTIVE

POSTOP COMPLICATIONS:

NIL

RECURRENCE

ANGULAR DEFORMITIES

PHYSEAL DAMAGE

GROWTH ARREST

INFECTIONS

REFRACTURE

IMPLANT LOOSENING

PATIE			LOCAT		FOLLO HEAL				PATH		ACTIVITY OF	
NT	AGE	SEX		SURGERY	W UP IN MONT HS	ING GRA DE	PRIOR SUR	POSTOP COM			FRAC	CYST
NO			ION									
1	14	F	F	NAIL AND BONE SUBS	36+	2	NIL	NIL			N	NO
2	9	M	F	NAIL AND BONE SUBS	24	2	NIL	IMPLANT LOOSENING			Y	YES
3	12	M	F	NAIL AND BONE SUBS	36+	2	NIL	RECURRENCE,INFECTION			N	YES
4	13	M	H	NAIL AND BONE SUBS	24	2	NIL	RECURRENCE			Y	YES
5	13	M	H	NAIL AND BONE SUBS	3	3	NIL	NIL			Y	NO
6	7	M	H	NAIL AND BONE SUBS	3	1	NIL	NIL			Y	YES
7	18	M	H	NAIL AND BONE SUBS	6	1	NIL	NIL			Y	NO
8	13	M	H	NAIL AND BONE SUBS	24	1	NIL	NIL			N	NO
9	3	M	H	NAIL AND BONE SUBS	36	1	NIL	NIL			N	YES
10	4	F	F	NAIL AND BONE SUBS	24	2	NIL	NIL			N	YES
11	10	F	H	NAIL AND BONE SUBS	6	1	NIL	NIL			N	YES
12	6	F	H	NAIL AND BONE SUBS	24	2	CURR AND BG	NIL			Y	NO
13	7	M	H	NAIL ONLY	12	1	CURR AND BG	NIL			Y	YES
14	9	F	H	NAIL AND BONE SUBS	24	1	NIL	NIL			Y	YES
15	12	M	H	NAIL AND BONE SUSB	12	1	NIL	NIL			Y	YES
16	6	M	H	NAIL AND BONE SUBS	12	1	NIL	NIL			Y	YES
17	8	F	F	NAIL AND BONE SUBS	18	1	CURR AND BG	NIL			N	YES
18	10	M	H	NAIL AND BONE SUBS	12	1	NIL	NIL			Y	YES
19	9	M	H	NAIL AND BONE SUBS	18	1	NIL	NIL			Y	YES
20	9	F	H	NAIL AND BONE SUBS	12	1	OTHERS	NIL			Y	YES
21	10	M	H	NAIL AND BONE SUBS	12	3	NIL	RECURRENCE			Y	NO
22	14	M	H	NAIL AND BONE SUBS	3	1	NIL	NIL			Y	YES
23	10	M	F	NAIL AND BONE SUBS	12	1	CURR AND BG	NIL			Y	YES
24	8	M	H	NAIL AND BONE SUBS	12	1	NIL	NIL			Y	NO
25	13	M	H	NAIL AND BONE SUBS	3	1	NIL	NIL			Y	YES
26	9	F	H	NAIL ONLY	6	1	NIL	NIL			Y	NO
27	12	M	H	NAIL AND BONE SUBS	6	2	NIL	NIL			Y	NO